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The prevalence of tracheal bronchus in pediatric patients undergoing rigid bronchoscopy

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Abstract: **BACKGROUND:** Tracheal bronchus (TB) is defined as an abnormal bronchus that originates directly from the lateral wall of the trachea above the carina and goes towards the upper lobe territory of the lung. We analyzed rigid endoscopies of the trachea in children to determine the incidence and characteristics of TB. **METHODS:** In total, 1021 rigid endoscopies of the trachea recorded from children aged 0 to 6 years were analyzed. Endoscopic examination was performed from supraglottic region to carina using a 0-degree Hopkins rod-lens telescope. Patients with a TB were identified and the site of origin of the TB and its level above the carina was noted. Data of the identified patients was reviewed for the presence of preoperative airway findings such as stridor, upper lobe pneumonia and wheezing or atelectasis, other congenital anomalies, and intraoperative complications. **RESULTS:** TB was detected in 11 (1.06%) of 1021 upper airway endoscopic examinations. All originated from the right lateral wall of the trachea. Six children had retained secretions in the TB, and 3 children had perioperative airway problems unrelated to the TB. One child showed right main stem bronchus narrowing as seen at the true carina, in the presence of a TB. All the children with TB exhibited at least 1 additional congenital anomaly at birth besides TB. **CONCLUSIONS:** TB is a relatively common congenital endoscopic lower airway anomaly in childhood, which is itself rarely symptomatic, but almost always coexists with other congenital anomalies.

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The Prevalence of Tracheal Bronchus in Pediatric Patients Undergoing Rigid Bronchoscopy

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Background: Tracheal bronchus (TB) is defined as an abnormal bronchus that originates directly from the lateral wall of the trachea above the carina and goes towards the upper lobe territory of the lung. We analyzed rigid endoscopies of the trachea in children to determine the incidence and characteristics of TB.

Methods: In total, 1021 rigid endoscopies of the trachea recorded from children aged 0 to 6 years were analyzed. Endoscopic examination was performed from supraglottic region to carina using a 0-degree Hopkins rod-lens telescope. Patients with a TB were identified and the site of origin of the TB and its level above the carina was noted. Data of the identified patients was reviewed for the presence of preoperative airway findings such as stridor, upper lobe pneumonia and wheezing or atelectasis, other congenital anomalies, and intraoperative complications.

Results: TB was detected in 11 (1.06%) of 1021 upper airway endoscopic examinations. All originated from the right lateral wall of the trachea. Six children had retained secretions in the TB, and 3 children had perioperative airway problems unrelated to the TB. One child showed right main stem bronchus narrowing as seen at the true carina, in the presence of a TB. All the children with TB exhibited at least 1 additional congenital anomaly at birth besides TB.

Conclusions: TB is a relatively common congenital endoscopic lower airway anomaly in childhood, which is itself rarely symptomatic, but almost always coexists with other congenital anomalies.

Key Words: tracheal bronchus, pediatric, bronchoscopy

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Tracheal bronchus (TB) was first described by Kubik and Muntener¹ in 1785 as a right upper lobe bronchus originating from the trachea. True TB is a congenital anomaly in which a right upper lobe bronchus has its origin in the trachea rather than at the carina. *Bronchus suis* and “pig bronchus” are alternative names used because a TB is a normal entity in swine.²

Demonstration of TB by computed tomography has been occasionally reported but only in isolated case reports.^{3–4} TB is associated with increased risk of pneumonia, atelectasis, air trapping and intubation, or weaning difficulties.^{5–6}

TB usually arises from the right lateral wall of the trachea (true TB) < 2 cm above the carina and can supply the entire upper lobe or just its apical segment, but it has been stated anecdotal as arising from the left side of the trachea also.⁷ A TB is said to be either *displaced* (if the anatomic upper lobe bronchus or its single branch is missing) or *supernumerary* (if the right upper lobe bronchus and its trifurcation is normal).⁷

Arbitrarily, TB has been classified into 3 types in the current literature (Figs. 1A–C).⁸ Type I is usually described to originate roughly at the junction of the middle and lower one third of the trachea. Type II is a small but distinct bronchus connected to the lower third of the trachea and type III is the TB arising from the tracheal wall almost at the level of the carina giving the appearance of trifurcated carina.

To date, only a few studies exist regarding the incidence of TB in humans.^{7,9}

Data are only available for the endoscopic presence of a TB and any associated comorbidities. The aim of this study is to retrospectively review > 1000 records of rigid endoscopies of the trachea for the prevalence, localization, and clinical presentation of TB in young children.

MATERIALS AND METHODS

A database of 1021 records of rigid endoscopies of the pediatric airway was used to analyze the incidence and characteristics of TB.

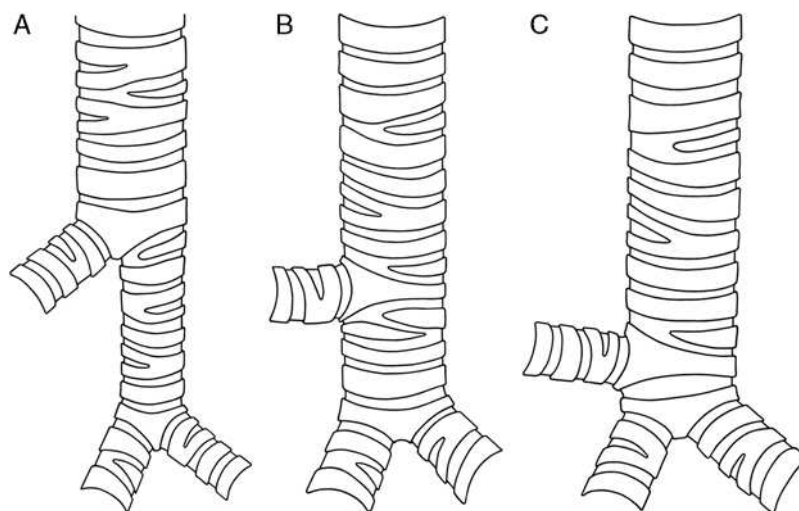


FIGURE 1. Schematic drawings representing tracheal bronchus types I (A), II (B), and III (C).

The database was created from an earlier prospective, observational clinical study performed at the Department of Anesthesiology, University Children's Hospital, Zurich, Switzerland from October 2008 to April 2011. This initial prospective study was aimed at investigating airway injuries in children (aged 0 to 6 y) with prior tracheal intubation compared with those without prior intubation and was performed with Ethical Committee Approval (KEK StV 24/08) and written parental consent. The results of this study are published elsewhere.¹⁰

Inclusion criteria in the former study were elective general anesthesia with planned tracheal intubation and muscle paralysis.¹⁰ Exclusion criteria were a known difficultly or abnormal airway, actual stridor, emergency procedures, risk of gastric regurgitation and patients of ASA class >III. Rigid tracheoscopy was performed using a folding anesthetic laryngoscope and Hopkins rod-lens telescope (0 degrees, 3.0 mm; Karl Storz 7200A, 7200 BW, 7200 D, GmbH, Germany) from supraglottis to carina and electronically recorded for later analysis. Rigid laryngo-tracheoscopies were performed by one of the 2 investigators (Weiss/Gerber) blinded to patient history and not involved in the patient's anesthetic management.

In the retrospective analysis of the 1021 endoscopic records of the trachea, patients with a TB were identified and the site of origin of TB and its level above the carina was noted. Preoperative data of the identified patients was reviewed to assess the presence of respiratory findings such as stridor, upper lobe pneumonia,

wheezing or atelectasis, and also the presence of associated congenital anomalies and/or syndromes. Intraoperative airway complications such as retained secretions, ventilation, and/or oxygenation problems as well as postoperative respiratory complications were noted from the patient's anesthesia and postanesthesia care unit records.

RESULTS

A TB was detected in 11 (1.06%) of the 1021 patients aged from 0 to 6 years (median age - 3 yrs). The TB arose from the right side of the trachea in all 11 patients. Reanalysis of the endoscopic video records confirmed an anomalous bronchus arising from the right lateral wall of the trachea above the carina (Fig. 2).

The TB in all these patients was of type III (Fig. 1C), which is a TB originating very close to the right main stem bronchus in the lower third of the trachea, within 2 to 4 cm above the carina, creating a typical trifurcating true carina as described in the literature.⁸

The clinical data of these 11 patients is shown in Table 1. The prevalence of TB was higher in females than in males (72%) (χ^2 test, $P = 0.01$). Seven of these 11 children had received previous multiple uneventful intubation anesthetics before the diagnosis of a TB was made in this study. Three children suffered from VACTERL complex with tracheoesophageal fistula, cardiac, and other congenital anomalies. Interestingly, in 3 otherwise healthy children undergoing excision of a congenital melanocytic nevus from the head or neck presented a TB was found as an additional solitary anomaly.

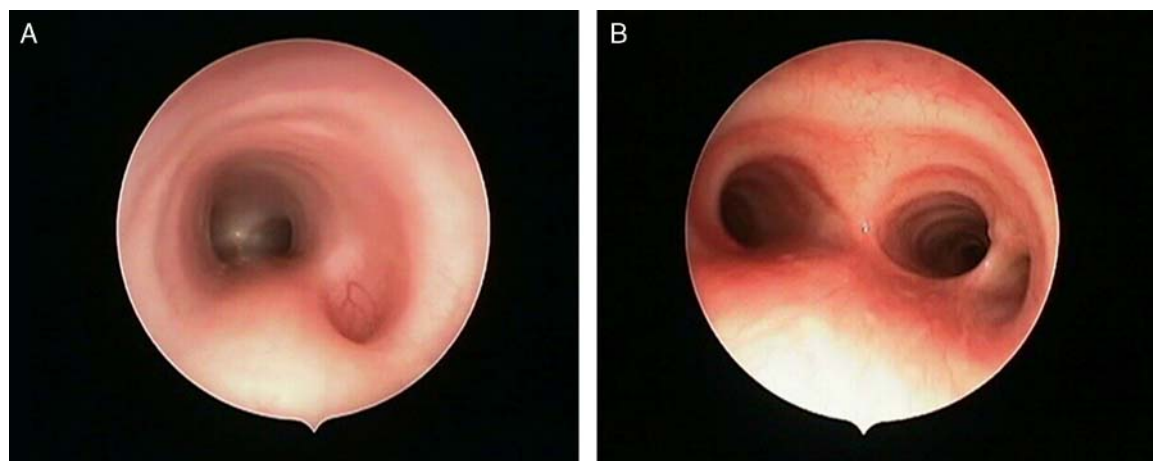


FIGURE 2. Hopkins rod-lens telescope view at the true carina. A, patient 7 (girl, 0.2 y); (B) patient 8 (boy, 5.5 y). Patient 1 (girl, 2.6 y); patient 2 (girl, 4.4 y); patient 3 (boy, 1.6 y); patient 4 (girl, 0.6 y); patient 5 (girl, 0.5 y); patient 6 (boy, 0.7 y); patient 7 (girl, 0.2 y); patient 8 (boy, 5.5 y); patient 9 (girl, 3.2 y); patient 10 (girl, 5.1 y); and patient 11 (girl, 1.8 y). *a+*

All children with a TB were in a stable medical condition, did not show respiratory symptoms, and did not require respiratory treatment before induction of anesthesia. Six children had retained secretions in the TB. One child showed right main stem bronchial narrowing or stenosis as seen at the true carina, in the presence of a TB. Three children had airway problems intraoperatively or postoperatively: 1 child with preintubation laryngospasm, perhaps due to stimulation in a light plane of anesthesia; 1 child with postoperative stridor requiring medical management; and 1 child with postoperative emergency delirium requiring medical treatment. All were unrelated to the TB. All patients with TB could be discharged from the postanesthesia care unit as planned, without respiratory complications that could be associated to the presence of an anomalous TB.

DISCUSSION

This study evaluated the prevalence, location, and related clinical manifestations as well as intraoperative and postoperative complications of TB in young children. The main finding was that TB is a relatively common but preoperatively asymptomatic endoscopic lower airway congenital anomaly, which always coexists with other birth defects.

We report an incidence of TB as 1.06% in our study which complies with that reported in the literature (0.1% to 2%).^{7,9} TB is often complicated by other congenital airway anomalies including laryngomalacia, tracheomalacia,

tracheal stenosis, and congenital heart disease.^{11–15} It is not surprising that all of the 11 children in our study had at least one other congenital anomaly at birth besides the presence of a TB. Three of these 11 children were born with congenital melanocytic nevus at birth and were posted for nevus excision in otherwise normal health state. To our knowledge, there exists no information in the literature associating the presence of TB to melanocytic nevus. The higher prevalence of TB in females (72%) than in males is another new finding of this investigation previously not reported. However, this will not have a significant clinical impact in day-to-day practice. Special attention should also be given to the fact that while in other studies, bronchoscopic examination was mostly carried out to examine respiratory pathologies or symptoms, in our study, majority of the patients were asymptomatic, admitted to the hospital for elective routine surgery other than airway or respiratory tract.

Right TB is reported more commonly in the literature than left TB.⁷ We observed only right-sided tracheal bronchi in our study population with the origin in the lower third of the trachea, within 2 to 4 cm above the tracheal bifurcation. This corresponds to the typical trifurcated appearance of the carina (type III) described by Conacher⁸ in adult patients.

There is anecdotal evidence of a real left TB^{16–18} in the literature. A prevalence of 0.3% to 1% for left TB has been reported in studies.^{7,9} However, attention should be paid to the fact

TABLE 1. Clinical Data of 11 Children With a Tracheal Bronchus as an Incidental Finding

Patients	Age (mo)	Sex	Associated Congenital Anomaly	Operations	Complications, Intervention	General Condition	Site of Origin	Right Main Bronchial Stenosis
1	31.2	F	VACTERL association congenital heart defect tracheoesophageal fistula spinal deformity, anterior anus hand and foot syndactyly	Hand surgery	None	Compromised	Right tracheal wall	No
2	52.8	F	Growth retardation congenital heart defect Ellis van Creveld syndrome hexadactyly both hands and feet anterior anus	Tibia osteotomy	None	Compromised	Right tracheal wall	No
3	19.2	M	Hypospadias	Correction of hypospadias	Postoperative breathing difficulty due to secretions requiring salbutamol inhalation	Healthy	Right tracheal wall	No
4	7.2	F	VACTERL association congenital heart defect high anal atresia, rectovesical fistula requiring colostomy absent coccyx, 11 ribs bilaterally vesicoureteric reflux	Cystoscopy	None	Compromised	Right tracheal wall	No
5	6	F	Preauricular accessory appendage	Excision of preauricular appendage	None	Healthy	Right tracheal wall	No
6	8.4	M	Hypospadias	Correction of hypospadias	Secretions that required tracheal suction. Postoperative airway problems requiring salbutamol inhalation	Healthy	Right tracheal wall	Yes
7	2.4	F	VACTERL association congenital heart defect high anal atresia requiring colostomy cleft palate, retrognathia, hand syndactyly	PSARP colostomy closure	None	Compromised	Right tracheal wall	No
8	66	M	Congenital melanocytic nevus of neck	Excision of nevus	None	Healthy	Right tracheal wall	No
9	38.4	F	Dandy Walker malformation vesicoureteral reflux psychomotor developmental abnormality limb spasticity, facial dysmorphism	Cystoscopy	Secretions, tracheal suction necessary	Compromised	Right tracheal wall	No
10	61.2	F	Congenital melanocytic nevus of cheeks	Excision of nevus	None	Healthy	Right tracheal wall	No
11	21.6	F	Congenital melanocytic nevus of forehead	Excision of nevus	None	Healthy	Right tracheal wall	No

PSARP indicates posterior sagittal anorectal plasty; VACTERL association (VATER syndrome), co-occurrence of birth defects Vertebral anomalies, Anal atresia, Cardiac defects, Tracheo-oesophageal fistula, Renal anomalies and Limb defects.

that most of the isolated case reports often have used different definitions of left TB. Erroneously, in these studies abnormal bronchi originating from the left main stem bronchus or from the apical, anterior or posterior upper lobe segment rather than from the trachea were included.^{16–18}

Type I anomalous TB (arising in the mid-trachea) might present a confusing appearance to a bronchoscopist undertaking foreign body removal or performing bronchoalveolar lavage. Likewise, it is easy to envisage many possible complications arising from endotracheal intubation in these cases. Either the TB or the true trachea could be entered by an endotracheal tube of normal length. While cannulation of the TB would obstruct most of the ventilation and pose a risk of pneumothorax, cannulation of the true trachea might bypass the right upper lobe and contribute to perioperative hypoxemia, atelectasis, or weaning difficulties in the intensive care unit.^{8,19–22} Use of short endotracheal tubes and fiberoptic confirmation of the tube position are of critical importance in such circumstances.

Type II and III anomalous TB is more likely to complicate the insertion of a double lumen tube (DLT) for single-lung ventilation. Stene et al²³ described the experience of intubating a patient with undiagnosed TB with a DLT leading to an unexpected combination of auscultatory and bronchoscopic findings, which were confusing for the anesthetist and dangerous for the patient. Conacher's group reported at least 3 cases in which an undiagnosed TB led to intubation or ventilation difficulty during attempted single-lung ventilation techniques. These authors advocate a diagnostic bronchoscopy in all patients requiring single-lung ventilation to avoid the hazard of lobe rupture or injury through the blind insertion of a DLT. Where a diagnosis of TB is confirmed, the literature suggests that use of a normal endotracheal tube and a bronchial blocker may be better than a DLT.⁸

However, flexible bronchoscopy (which invariably confirms the diagnosis of TB) was not performed in this study. Any other coexisting lower airway anomalies were not examined to determine whether the right accessory lobe bronchus actually ventilated some part of the lung or was just a blind pouch.

As in the presented study, patients with a TB are usually asymptomatic and diagnosed incidentally by bronchoscopy or chest computed tomography for other respiratory diseases.

Recent reports indicate that an increasing number of patients with TB present with wheezing, stridor, cough,¹⁵ or sometimes even recurrent episodes of infection (due to retained secretions), hemoptysis, or malignancy.^{24–26} Treatment of TB is based upon the severity of symptoms. Most patients with a TB can be managed conservatively. However, when respiratory problems such as recurrent pneumonia, atelectasis, or air trapping persist, surgical excision of the involved segment may become necessary.²²

TB is not a rare finding but it is rarely symptomatic, and hence its undiagnosed presence can be crucial in certain special situations such as foreign body aspiration and single-lung ventilation. Patients with unexplained oxygenation problems during endotracheal intubation and patients with recurrent respiratory infections should alert the treating physician to the possibility of a TB. Apart from such special situations, TB exists innocently in the body, sometimes requiring conservative management but rarely needing surgical intervention.

CONCLUSIONS

TB is a relatively common endoscopic lower airway congenital anomaly in childhood, which is itself rarely symptomatic, but almost always coexists with other birth defects.

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